

A Rare Case of Synchronous Squamous Cell Carcinoma on the Body of the Tongue and Malignant Lymphoma of the Neck

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Abstract

Synchronous malignancy with squamous cell carcinoma (SCC) in the head and neck occurs mainly in the aerodigestive tract, with a frequency of 60-70%. In contrast, SCC in the head and neck very rarely coincides with malignant lymphoma (ML). Here we report a case of synchronous SCC on the lower-left surface of the tongue and ML on the left -side of the neck in a 75-year-old man. The patient had consulted a local doctor for a mass on the left side of his neck. After enucleation of the neck mass, the pathological diagnosis was metastatic malignant tumor of unspecified origin. On the other hand, the tongue lesion was regarded as benign and followed-up. For further diagnosis of the lesions, the patient was transferred to our hospital in March 2015. Biopsy indicated tongue SCC. Resection of the primary tongue lesion and radical neck dissection of the left side neck were then performed. The cervical mass was diagnosed as ML, and found to be simultaneously affected by the tongue SCC and cervical ML. Approximately two years later, a follow-up CT showed recurrent ML on the left tonsil, which was treated by chemotherapy.

Keywords

Synchronous neoplasms; Squamous cell carcinoma; Malignant lymphoma; Head and neck

Introduction

The incidence rate of synchronous carcinoma is approximately 11.0 ~ 16.2% [1]. The second primary malignancy in patients with head and neck carcinomas is usually located in the head and neck region (40%), lung (31%), or esophagus (9%) [2]. However, the simultaneous occurrence of tongue squamous cell carcinoma (SCC) and B cell lymphoma in the neck is extremely rare. To our knowledge, only three cases of synchronous cancer involving SCC of the tongue and cervical ML are reported in the English literature, and in those reports, the SCC lesion was on the base of the tongue [3,4]. Furthermore, double cancer involving SCC on the body of the tongue and jugular ML is thought to be very rare. Here, we report a case of synchronous malignancy consisting of SCC located on the body of the tongue and B-cell lymphoma presenting as a neck mass.

Case Presentation

A 75-year-old man who was fully active became aware of pain on his tongue and a painless mass on the left side of his neck in January 2015. At the initial hospital, a white lesion on the lower-left surface of the tongue was regarded as benign and subjected to follow-up observation, whereas an operation of the cervical mass led to a pathological diagnosis of metastatic malignant tumor of unspecified origin. To determine the relationship, if any, between the white lesion of the tongue and the metastatic tumor in the neck, and for further treatment, the patient was admitted to our hospital in March 2015.

Extraoral examination revealed an elastic soft and mobile mass without tenderness on the left side of the cervical region, the size of a thumb tip, based on palpation (Figure 1a). Intraoral examination revealed a white lesion, not surrounded by in duration, on the lower left surface of the tongue, 21 mm × 15 mm in size (Figure 1b). We then performed a biopsy of the tongue lesion. The pathological diagnosis of this lesion indicated tongue SCC, based on the presence of multiple proliferating deformed and acidophilic heterocysts, with parakeratinized stratified squamous epithelium, intercellular bridges, and keratin pearls (Figure 2).

Contrast-enhanced magnetic resonance imaging (contrast-enhanced MRI) and

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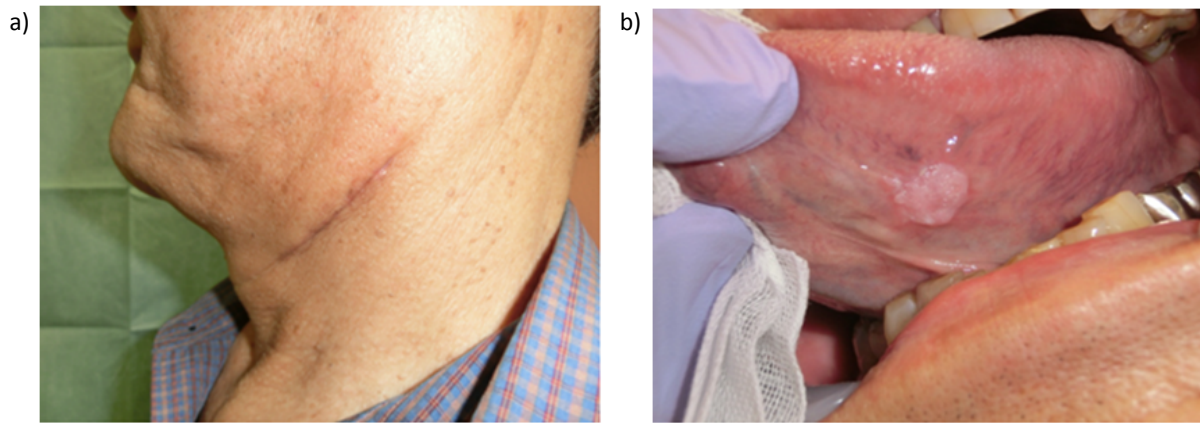


Figure 1: Clinical examination. (a) Extraoral view showing a slightly elevated mass near the left cervix. (b) Intraoral view revealing a regularly white lesion on the left lower surface of the tongue.

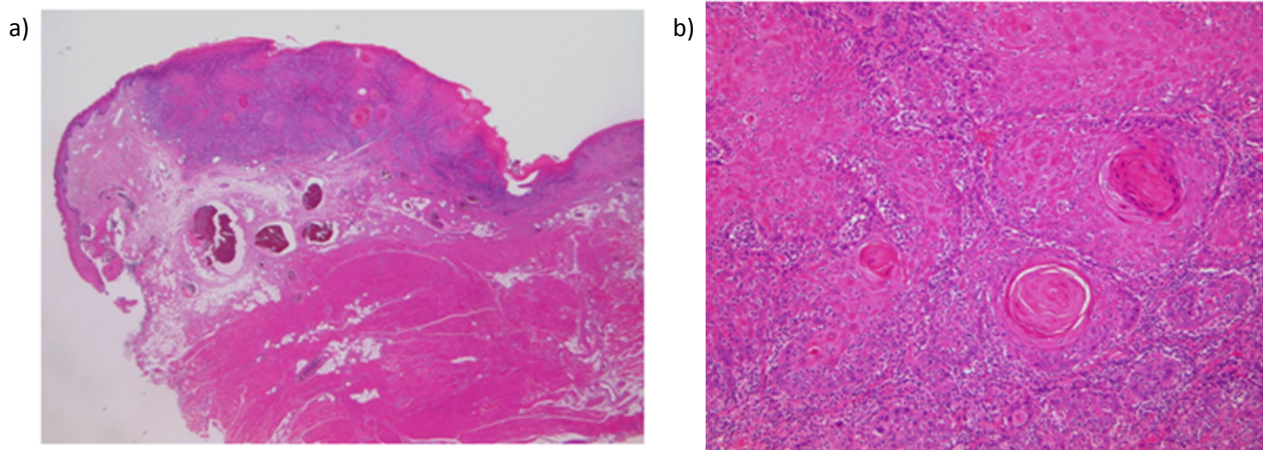


Figure 2: Pathological diagnosis based on biopsy of the tongue lesion. Panel (a) shows the multiple proliferating deformed and acidophilic heterocysts with para keratinized stratified squamous epithelium (original magnification x 12.5). Panel (b) shows intercellular bridges and keratin pearls among the cysts (original magnification x100).

18F-Fluoro-deoxy-glucose positron emission tomography (FDG-PET/CT) scans were performed. Contrast-enhanced MRI (T1-weighted images) revealed a lesion on the tongue measuring about 12 mm × 12 mm with a depth of almost 2 mm, indicating poor invasion. The tongue lesion was slightly enhanced in the T1-weighted images after gadolinium administration (Figure 3a). The scans also showed an approximately 30 mm × 25 mm mass in the back-left region of the submandibular gland (Figure 3b). No swelling was observed in the other lymph nodes. The FDG-PET/CT scan also detected a region of sharply enhanced FDG uptake (SUV max=17.9) with a maximum diameter of 23.5 mm in the left cervical field (Figure 3c). In contrast, the tongue cancer could not be visualized by FDG-PET/CT, which is unusual for metastatic SCC. Based on these results, we diagnosed this case as tongue SCC T1N2bM0 by UICC/AJCC staging.

Therefore, we considered the possibility of tongue SCC with metastatic lymph nodes. In April 2015, under general anesthesia, a partial glossectomy and modified radical neck dissection (mRND) were performed. The tongue specimen did not include residual SCC. This may be the reason why the SCC had already been removed from the tongue when we performed biopsy of the tongue lesion. The pathological examination revealed that the cervical lymph node did not include malignant tumor cells derived from the tongue lesion, but had features of B-cell lymphoma (Figures 4a,4b). An

immunophenotypic study of the neck specimen, classified it as B-cell lymphoma (Figures 5a,5b,5c). Immunohistochemistry showed that the tumor was positive for CD20, bcl-2, Ki-67, CD79a, and CD21, and negative for CD3, CD5, CD30, CD10, cyclinD1, and bcl-6. Due to inadequate fixation of the specimen, categorization of the B-cell lymphoma was difficult, although follicular lymphoma was suspected as the most likely diagnosis.

The stage of the B-cell lymphoma was Ann Arbor. Thus, the diagnosis was synchronous carcinoma with tongue SCC and B-cell lymphoma.

The patient was subsequently followed up for signs of recurrence in the left cervix and metastases of the tongue by the Hematology department and our department. In September 2017, although recurrence of the tongue cancer has not been detected, malignant lymphoma in the palatine tonsilla was detected by follow-up CT scans, and the patient is receiving chemotherapy with R-Bendamustine once a month.

Discussion

In cancer of the oral region, it is not unusual to encounter a case of tongue SCC and cervical mass. In most of these cases, the neck mass is diagnosed as a metastasis from the oral SCC. However, neck

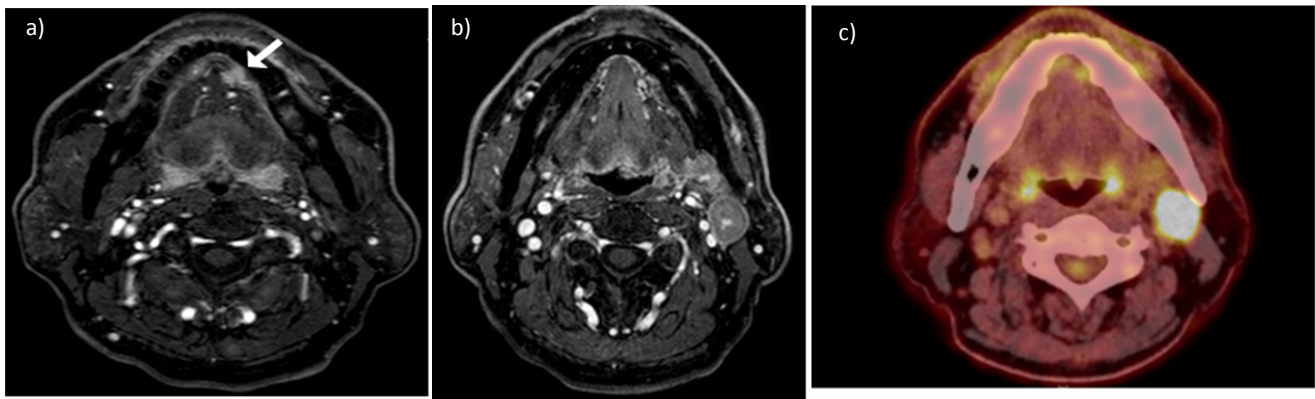


Figure 3: Contrast-enhanced axial T1-weighted MR. (a) The tongue lesion was approximately 12mm×12mm, and its depth was almost 2mm. (b) MR image of the neck revealing an approximately 30 mm × 25 mm mass. (c) FDG-PET/CT image showing an FDG-positive mass located in Level II

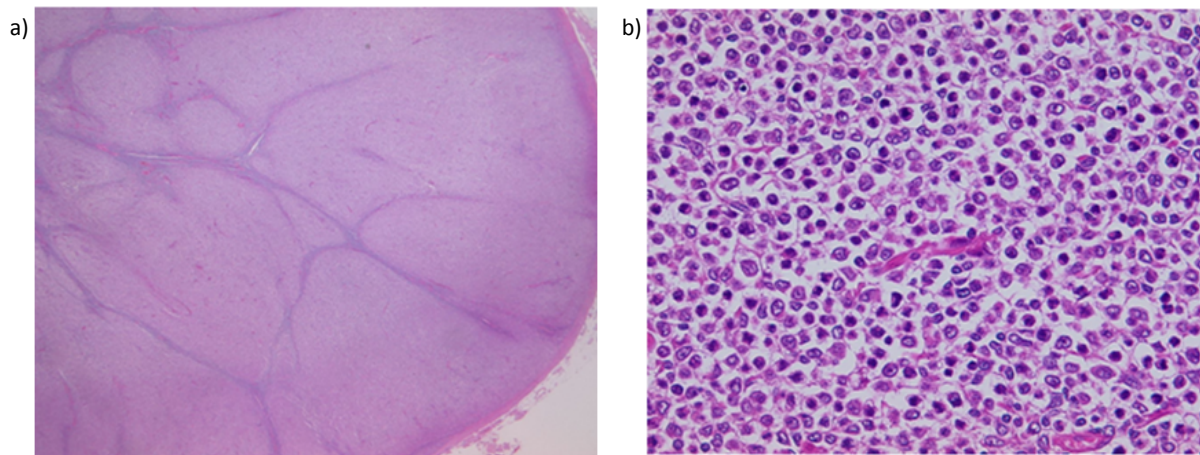


Figure 4: Photomicrograph of the mass in the left side of the neck. (a) Low-power view revealing the histology of some sort of B-cell lymphoma. (b) Higher magnification showing irregular nodular and proliferating atypical lymphocytes of small to moderate size.

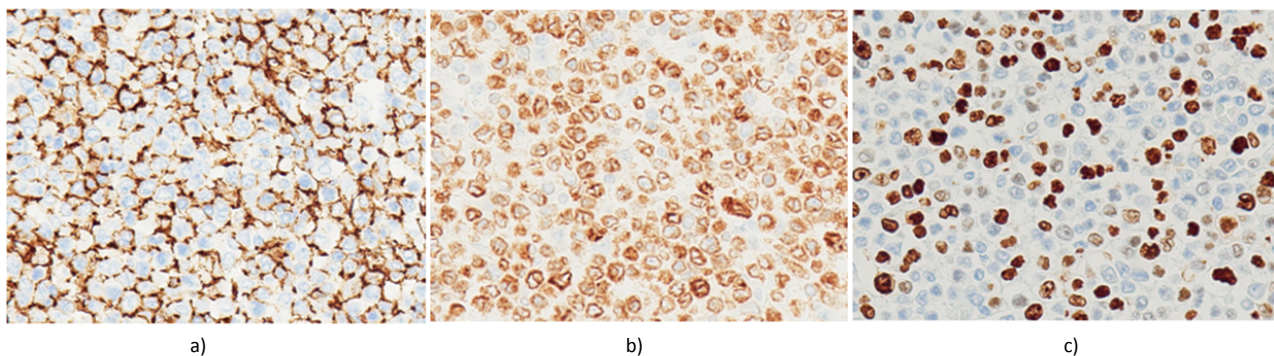


Figure 5: Immuno phenotypic study of the neck specimen: Images show positive immune staining for (a) CD20, (b) Bcl-2 and (c) 40%-50% Ki-67 (original magnification x 400).

dissection can reveal unexpected findings, as in our case. Sheahan et al. performed a retrospective review of 202 patients and concluded that unanticipated findings are occasionally encountered during the pathologic examination of neck dissections [5]. Specifically, unpredicted pathologies were discovered during the neck dissection in more than 5% of patients with head and neck carcinoma, nearly half of which were malignant tumors [5]. In our case, the simultaneous

carcinoma consisting of tongue SCC and cervical ML fits the definition of unexpected findings and represents an unusual case.

It is well known that most second primaries in head and neck carcinoma patients occur in the lung, head and neck, or esophagus. Leon et al. noted that the second primary malignancy in patients with head and neck carcinomas was predominately situated in the head

and neck (40%), lung (31%), or esophagus (9%) [2]. Furthermore, when simultaneous primaries occur in the head and neck, the coincidence of SCC and ML appears to be very rare. Watanabe et al. reported that simultaneous malignancies of SCC and ML in the head and neck region are extremely uncommon; they only knew of two cases in the literature [6]. Consistent with that study, in Sheahan et al.'s survey of 202 consecutive patients with SCC in the head and neck, only ten patients had unexpected findings, only one of which was lymphoma [5]. Similarly, Young-Hoo et al reported that to their knowledge, there were ten cases of multiple primary malignancies with head and neck SCC and malignant lymphoma [7].

Moreover, in the co-occurrence of SCC and ML, tongue SCC with cervical ML-like our case is thought to be extraordinarily uncommon. Watanabe et al. reported that SCC of the skin appears to be the most common second primary malignancy in patients with ML [6]. Moreover, Sheahan et al. reported that only one case of skin SCC with Non-Hodgkin's Lymphoma (NHL) was detected among 274 patients with SCC who were retrospectively reviewed [5]. Thus, it appears that even double cancers with skin SCC and NHL seldom occur, and are even rarer when the SCC and ML are at other anatomic sites, for example, tongue SCC and neck ML. Furthermore, Stack et al reported that only 2 cases of synchronous carcinoma involving SCC on the base of the tongue and neck ML were found among 866 patients diagnosed with SCC from 1988 to 1993 at their institutions [3]. Similarly, a case of double cancer involving SCC at the base of the tongue and neck ML was deemed unusual by Millwaters et al [4]. The tongue base is adjacent to the pharynx, and ML of the tongue base cannot be easily distinguished from pharyngeal ML. Thus, the simultaneous occurrence of SCC at the body of the tongue and cervical ML, as observed in our case, appears to be extremely rare. In fact, to our best knowledge, there are no reports of SCC on the body of the tongue with cervical ML.

In terms of the elevated risk of Second Primary Malignancy (SPM), it is notable that patients with head and neck SCC have a significantly elevated risk of SPM, most commonly within the head and neck, lung, or esophagus. Morris et al. reported that in patients with index head and neck SCC, the risk of lung cancer was most markedly elevated, followed by head and neck and esophageal cancer, whereas a smaller but significant increase in the risk of lymphoma was found [8]. Furthermore, when the index SCC was in the oropharynx or larynx, the increased risk of lymphoma was slightly elevated, but it was not elevated when the index SCC was in the oral cavity, including the tongue [8]. These results were based on research conducted by the National Cancer Institute's Surveillance, Epidemiology, and End Results (SEER) program, the registry of which contains a total of 75,087 cases identified between 1975 and 2006 [8]. In summary, this population-based cohort study showed that head and neck SCC patients have an elevated risk of SPM in the lung and bronchus, head and neck, or esophagus, and a significantly elevated but lower risk of SPM as lymphoma [8]. Accordingly, from the viewpoint of elevated risk, the synchronous occurrence of tongue SCC and cervical ML, as in our case, appears to be very rare.

ML is broadly classified into Hodgkin Lymphoma (HL) and Non-Hodgkin Lymphoma (NHL), and the number of NHL cases, which include follicular lymphoma and small cell lymphocytic lymphoma, exceeds that of HL [3-6]. The B-cell lymphoma in our case was categorized as NHL; in Japan, NHL is much more common than HL, with frequencies of 90% and 7%, respectively [9].

Thus, it appears that the malignancies seen in our case (tongue SCC and cervical ML) seldom coexist. Nevertheless, because the coexistence of SCC and ML generally result in a poor prognosis, a cervical mass should be carefully diagnosed to guide treatment for the best prognosis. Gluckman and Crissman reported an overall 5-year disease survival rate of 35% for single cancer and 22.3% for multiple primary cancers [10]. Likewise, Boddie et al. reported that patients with reticuloendothelial malignancy and primary tumors of the oral cavity had a very short survival time (average, 8.5 months) [11]. Accordingly, it is important to examine a neck mass carefully as soon as it is detected. In this respect, ultrasonography (USG) and Fine Needle Aspiration (FNA) biopsy are useful for checking a cervical

mass. According to Joshi, USG has many advantages, including being radiation free, broadly available, simple to use, noninvasive, and inexpensive[12]. Furthermore, according to Ying et al., the cervical lymph nodes are commonly evaluated by CT and MRI, which are less effective than USG for identifying small nodes [13]. Ultrasound examination of the cervical lymph nodes should be routinely applied by clinicians to obtain a diagnosis without delay. USG is also used to guide FNA biopsy.

Considering that cases of double cancer have increased, when a cervical mass is found in a clinical examination, synchronous malignancy in the head and neck region should be suspected [14].

Conclusion

The simultaneous occurrence of tongue SCC and neck ML appears to be a very unusual event, especially the coexistence of SCC at the body of tongue and cervical ML, as in our case. Therefore, considering that such cases can still exist, it is important to obtain a precise diagnosis using USG and FNA as early as possible, to guide the treatment plan and improve the prognosis.

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